

Idiopathic Lingual Artery Aneurysm: CT Findings and Endovascular Therapy

A Case Report

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Summary

We describe a 65-year-old woman with an asymptomatic idiopathic lingual artery aneurysm which is suspected to be congenital. We review the literature on external carotid artery branch aneurysms, diagnostic evaluation and discuss treatment options for the various types and the specific chosen in the case presented.

Introduction

External carotid artery (ECA) branch aneurysms are extremely rare vascular abnormalities¹, usually occurring in the more superficial branches. Aneurysms of the lingual artery (LA) are particularly rare, usually pseudoaneurysms associated with trauma, neoplasms, surgery or radiofrequency ablation²⁻⁵. Only three cases of idiopathic, possibly congenital, aneurysms of the LA have been reported in the literature⁶⁻⁸. LA aneurysms can be fatal if not recognized and treated in a timely manner⁹. All of the reported histological analyzed LA aneurysms have been described as pseudoaneurysms. We describe the case of a 65-year-old woman with an asymptomatic LA aneurysm which was incidentally discovered on a CT of the neck obtained for presurgical evaluation for hyperparathyroidism. The aneurysm was confirmed by digital subtraction angiography (DSA) and successfully treated using platinum coils. We discuss presenting symptoms, etiologies, imaging findings and treatment options.

Case Report

A 65-year-old woman was evaluated presurgically for hyperparathyroidism and related hypercalcemia with a contrast-enhanced CT of the neck, which was performed for correlation with a previously acquired sestamibi scan. On the CT a 1.2 x 1.0 cm hyperdense vascular mass was identified at the left tongue base arising from the LA, with incomplete opacification of the outpouching suspicious for a partially thrombosed aneurysm (Figure 1A, B). Thorough scrutinization of the patient's history revealed no trauma, intervention, history of aneurysms, or malignancy to account for the aneurysm.

DSA was performed and a left-sided 13 mm LA aneurysm was confirmed (Figure 2). Endovascular obliteration of the aneurysm was offered considering the potential consequences of aneurysm rupture, particularly since endotracheal intubation was required for the patient's planned surgery. The aneurysm was successfully treated using platinum coils (Figure 3). The patient was discharged without complication.

Discussion

External carotid artery (ECA) aneurysms have long been known to comprise 2.2% of all cervical carotid aneurysms¹. Aneurysms of the superficial temporal and facial arteries are the most common, with aneurysms of the LA considerably more rare. Idiopathic aneurysms of

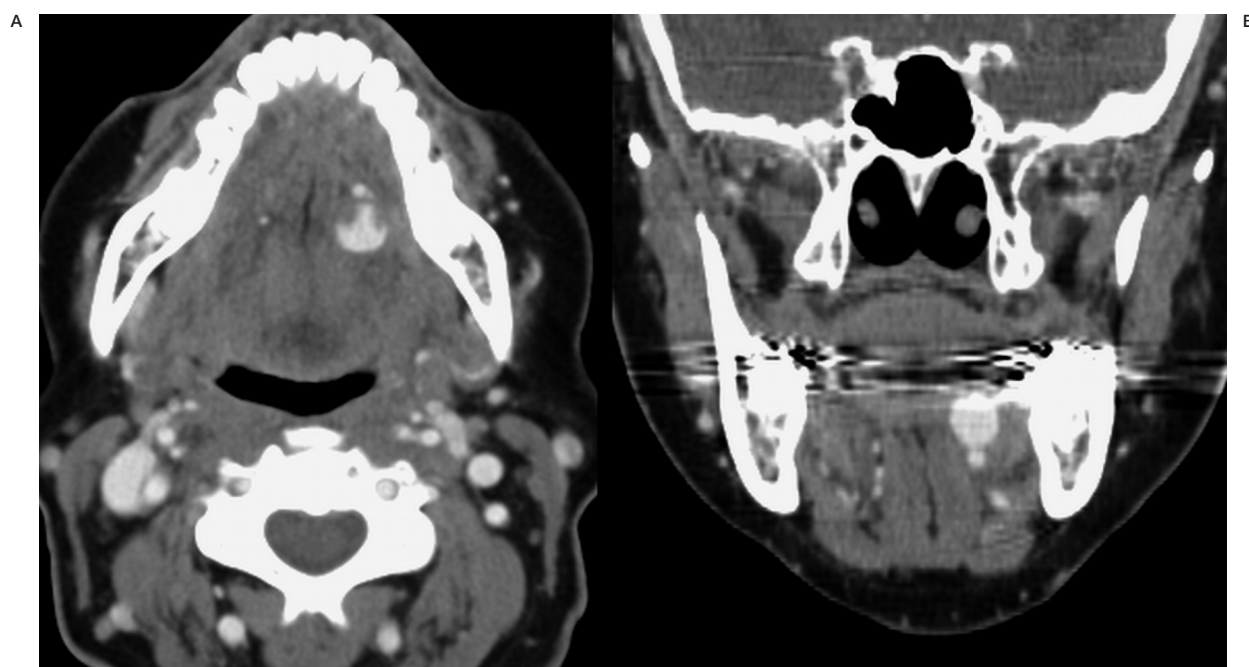


Figure 1 A) Contrast-enhanced axial CT at the level of the base of the tongue demonstrates a contrast-enhancing mass in the left base of the tongue with relative sparing of the anterior bilateral aspects concerning for a partially thrombosed LA aneurysm. B) Coronal reconstruction of the axial CT demonstrates the topographic relation to the LA.

the LA are especially rare, with only three reported cases in the English literature⁶⁻⁸. Two of these reported cases^{6,7} were surgically resected and found histologically to be most consistent with pseudoaneurysms. Both cases initially presented as pulsatile masses. Adib et Al reported

bilateral LA aneurysms, which were discovered only after one of the aneurysms ruptured. These are, to date, the only reported cases that are suggested to possibly represent congenital aneurysms.

The vast majority of ECA aneurysms are pseudoaneurysms with known etiology, mostly traumatic or postsurgical. Nearly all of the reported LA aneurysms are iatrogenic, in particular related to tonsillectomies^{4,10,11}. However, other interventions have also caused LA pseudoaneurysm formation such as radiofrequency tongue base reduction for obstructive sleep apnea⁵. Penetrating trauma is another less common, but well-known cause of LA aneurysm². Other reported causes of pseudoaneurysm formation of the LA include treated neoplasm with both surgery^{12,13} and radiation³, as well as odontogenic infection¹⁴.

Most of these aneurysms were discovered as a result of hemorrhage, with the need for urgent or emergent hemostasis^{3-5,12}. When hemorrhage is identified, as it is often the presenting symptom, rapid intervention is indicated as this is potentially life-threatening^{4,9}. Pseudoaneurysm formation occurs as a result of injury to the artery, and subsequent hematoma development connecting to the arterial lumen, with on-



Figure 2 Frontal projection DSA image shows selective injection of the left lingual artery aneurysm.

ly a fibrous tissue-lined cavity and pseudointima⁸. This is likely the reason for their predisposition to rupture. True aneurysms, on the other hand, demonstrate dilation of one or more of the native vascular layers, and the only reported case of a ruptured possible congenital true LA aneurysm resulted only in ecchymosis⁸. In our case endovascular therapy was utilized, and histologic evaluation of the aneurysm could not be performed to determine if this represented a true or false aneurysm.

As endovascular treatment techniques have evolved, these have become increasingly favored over surgical ligation of ECA branch aneurysms¹². Schroth et Al reported using platinum coils for a lingual artery aneurysm as early as 1991³. Since then coiling, gelfoam, and the use of liquid embolic agents such as n-butyl cyanoacrylate (NBCA) have been the treatment options most frequently employed^{5,10-12,14}, even in emergent cases⁴. In the case reported by Matsumoto et Al a small transient infarct in the left frontal lobe occurred within three hours of embolizing the LA pseudoaneurysm with NBCA, and they speculated that a small piece of adhesive may have embolized at the time of catheter removal¹².

In contrast to most of the reported cases of LA aneurysms, our patient had no history of trauma, malignancy, or surgical intervention to suggest a possible etiology, raising the suspicion that this could represent a congenital aneurysm. Even in contrast to the idiopathic LA aneurysms previously reported⁶⁻⁸, the patient in our case was entirely asymptomatic, and the aneurysm incidentally detected on a routine CT of the neck. The decision was reached to embolize the aneurysm pre-emptively using platinum coils, which were chosen over NBCA mainly because of the aneurysm size of 13 mm, which as a hard mass of NBCA could produce some discomfort to the patient. There have



Figure 3 Left external carotid artery injection after coiling demonstrates embolization of the left lingual artery aneurysm.

been reports on compromised vascular flow of distal aspects of the lingual artery, and thereby impairment of the motor and sensory function of the distal tongue as well as NBCA reflux into other ECA branches and even the internal carotid artery when rapidly retrieving the catheter¹².

In conclusion, LA aneurysms are rare vascular lesions, which in the majority are acquired. Congenital LA aneurysms have been demonstrated, but require histologic evaluation for confirmation, which is often not practical and with endovascular treatment has become impossible. Despite lacking a definitive etiology the radiological presentation and associated potential risks warranted prompt intervention.

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